

Matching in Prosopagnosia 1

The Domain-Specificity of Face Matching Impairments in 40 Cases of Developmental Prosopagnosia

Abstract

A prevailing debate in the psychological literature concerns the domain-specificity of the face recognition system, where evidence from typical and neurological participants has been interpreted as evidence that faces are “special”. Although several studies have investigated the same question in cases of developmental prosopagnosia, the vast majority of this evidence has recently been discounted due to methodological concerns. This leaves an uncomfortable void in the literature, restricting our understanding of the typical and atypical development of the face recognition system. The current study addressed this issue in 40 individuals with developmental prosopagnosia, completing a sequential same/different face and biological (hands) and non-biological (houses) object matching task, with upright and inverted conditions. Findings support domain-specific accounts of face-processing for both hands and houses: while significant correlations emerged between all the object categories, no condition correlated with performance in the upright faces condition. Further, a categorical analysis demonstrated that, when face matching was impaired, object matching skills were classically dissociated in six out of 15 individuals (four for both categories). These findings provide evidence about domain-specificity in developmental disorders of face recognition, and present a theoretically-driven means of partitioning developmental prosopagnosia.

Keywords: Developmental prosopagnosia; domain specificity; face recognition; face perception; visual agnosia.

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1. Introduction

Prosopagnosia is a visuocognitive condition that is characterized by an inability to recognise others by their face. It has traditionally been studied in individuals who acquire face recognition difficulties following neurological injury (acquired prosopagnosia: AP; Barton, 2008; Bate et al., 2015; Dalrymple et al., 2011; Damasio, Damasio, & Van Hoesen, 1982), but has more recently been documented in a larger number of people with developmental origins (developmental prosopagnosia: DP; Bennetts, Murray, Boyce & Bate, 2017; Bate et al., 2019; Bowles et al., 2009; Duchaine, 2008). Several investigations indicate that DP is a heterogeneous condition (e.g. Bate & Bennetts, 2015; Burns, et al., 2017; Le Grand et al., 2006; Minnebusch, Suchan, Ramon & Daum, 2007; Schmalzl, Palermo & Coltheart, 2008), although there has been little progress in establishing specific phenotypes. While the similarities between AP and DP are still being investigated, the former offers a broad yet theoretically-driven starting point for the partitioning of its developmental equivalent: the presence or absence of co-morbid impairments in object processing (e.g. De Renzi, 1986; Farah, Wilson, Drain, & Tanaka, 1995; McNeil & Warrington, 1993).

Much work has examined object recognition abilities in AP as a means to inform the long-standing debate regarding the domain-specificity of the face recognition system (Diamond & Carey, 1986; Gauthier & Logothetis, 2000; Kanwisher, 2017; McCarthy, Puce, Gore & Allison, 1996). While case studies of APs with preserved object recognition skills have supported modular accounts of functionally distinct cortical face and object representations (Busigny, Graf, Mayer & Rossion, 2010; Kanwisher, 2017; McCarthy et al., 1996; Sergent & Signoret, 1992), some suggest that those with more widespread impairments

in visual recognition imply modules that are specialized for any expert visual category (e.g. Gauthier, Behrmann & Tarr, 1999), or support domain-general hypotheses of distributed cortical function (e.g. Behrmann & Plaut, 2015; Haxby et al., 2001) and/or common underlying mechanisms for different object categories (e.g. sensitivity to curvature: Nasr, Echavarria & Tootell, 2014; Ponce, Hartmann & Livingstone, 2017; spatial frequency: Woodhead, Wise, Sereno & Leech, 2011; reliance on holistic processing: Richler, Palmeri & Gauthier, 2012).

Despite there being many more cases of DP available for investigation, progress has been slow. While individuals with preserved (Dobel, Bolte, Aicher & Schweinberger, 2007; Duchaine, Dingle, Butterworth & Nakayama, 2004) or impaired (Behrmann, Avidan, Marotta & Kimchi, 2005; Duchaine, Germine & Nakayama, 2007; Esins et al., 2016) object recognition skills have been reported, many studies only report accuracy data (but see Duchaine et al., 2004). Such work has been questioned by claims that seemingly normal object recognition performance may be underpinned by laboured or sub-optimal processing strategies (Gauthier, Behrmann & Tarr, 1999; but see Rossion, 2018; Starrfelt & Robotham, 2018, for discussion).

A second limitation of previous work on object agnosia in DP is that many studies have only examined one comparison object, making it difficult to determine whether the individual represents a true case of pure prosopagnosia without object agnosia. However, in a recent review of all DP cases reported to date, Geskin and Behrmann (2018) noted that when more than two object categories were included, no individual was classified as having intact object recognition. Unfortunately, this introduces a fundamental asymmetry to the literature: with a large number of comparisons (e.g. accuracy and reaction time for 3+ object categories) it is easy to identify - and possibly misidentify – a potential deficit in object recognition. For example, Garrido, Duchaine, and DeGutis (2018) noted that 50% of control participants from

a previous study could have been classified as having an object recognition impairment using Geskin and Behrmann's criteria. On the other hand, with so many potential object categories, it is also simple for authors to dismiss cases of ostensibly intact object recognition by claiming that the “wrong” objects have been studied.

Further, many authors offer little justification for choosing a particular object (e.g. Huis in't Veld, Van den Stock, & de Gelder, 2012; Shah, Gaule, Gaigg, Bird, & Cook, 2015; Zhao et al., 2016; but see Malsapina, Albonico, Toneatto, & Daini, 2017). This has led to a bewildering and inconsistent range of object tests being used, which can limit comparisons between studies. One solution is to treat all object categories as equally valid for identifying potential deficits. While useful for pointing to potential associations or dissociations between faces and objects on a broad level, this approach can make it difficult to draw wider conclusions about the perceptual or cognitive origins of the disorder. Rather than object processing as a whole being “intact” or “impaired” in DP, it is possible that a selective deficit is only apparent for some object classes – for example, bodies or body parts (e.g. AP case FM, Moro et al., 2012; DP cases in Righart & de Gelder, 2007); or that the deficit is present for any stimuli that share perceptual processing demands with faces, such as a first order configuration and within-category discrimination (e.g. AP patient LH, de Gelder & Rouw, 2000; DP cases in Malsapina et al., 2017). Consequently, it may be more theoretically informative to examine the pattern of deficits across different objects, as opposed to simply classifying a deficit as being present or absent.

These shortcomings are unfortunate given the larger prevalence of DP compared to AP should allow the co-occurrence of object agnosia to be more systematically examined. In the only large-scale empirical study reported to date, Zhao et al. (2016) reported a weak significant correlation between face and object (flowers, cars and birds) perception in 64 DPs, and later categorical analysis of the dataset concluded that 40 DPs had normal object

perception skills, seven had mild deficits, and 17 showed severe impairments (Geskin & Behrmann, 2018). However, the design and difficulty of the study were not matched across the face and object conditions: while the former used the faces of celebrities (thereby drawing on mechanisms used in familiar face recognition: Johnston & Edmonds, 2009), the object condition used unfamiliar exemplars for matching. Furthermore, the analysis did not discriminate between different types of objects, so it is not possible to determine whether the object recognition deficits were present across all categories, or whether different individuals showed different clusters of impairment.

The review reported by Geskin and Behrmann (2018) attempted to draw conclusions across all published studies, concluding that ~20% of DP cases are face-specific, and that the frequent association between face and object recognition supports a domain-general explanation of DP. However, this figure is likely to be inaccurate given that (a) over a third of cases were excluded as insufficient data were available, (b) there were vast differences across studies in the diagnostic procedures used to identify DPs, and (c) many studies failed to select appropriate object categories, resulting in variations in task difficulty between the face and object measure. In fact, when the large sample reported by Zhao and colleagues is dismissed, alongside another case where inappropriate methodology was used (Weiss, Mardo & Avidan, 2016; see Campbell & Tanaka, 2018), the face-specific category only contains six individuals (less than 1% of the sample; but see Garrido, Duchaine & DeGutis, 2018, for a potential omission). However, it is very difficult to draw strong conclusions across these remaining cases, given different sub-processes of object recognition were tapped in each study.

The current study assessed face- and object-processing skills in 40 DP participants, using a sequential same/different face and object (hands, houses) matching task. The identical paradigm was used across all three stimulus categories, presenting perceptually homogenous stimuli which required discrimination on an exemplar (as opposed to a category) level (see

Campbell & Tanaka, 2018). We selected the two object categories for theoretical reasons.

Hands share several perceptual characteristics with faces: they are frequently seen body parts that share a first-order configuration (i.e. a specific arrangement of features common to all specimens); and the variability in hands is genetically driven, as opposed to experimentally controlled (see Crookes & McKone, 2009). There has also been substantial interest in the relationship between face and body processing, with some suggestion that impairments in face-processing arise from cognitive and neural mechanisms that are shared with body perception (Righart & de Gelder, 2007; but see Biotti, Gray, & Cook, 2017). Pertinently, neural areas associated with body perception (the extrastriate body area: Downing, Jiang, Shuman, & Kanwisher, 2001) are responsive during the perception and discrimination of isolated hand stimuli (Myers & Sowden, 2008). Thus, if DPs demonstrate a deficit for body parts, or biological objects more generally, we would expect this to be present for hands. On a more practical level, hands are also commonly viewed in their natural state, whereas bodies are usually viewed clothed, which may affect the cues people use to recognise them.

Furthermore, we deemed hands to be preferable to other body-related stimuli such as biological motion (e.g. point-light walkers; Johansson, 1973), as movement may influence reaction time measures (e.g. people may not be able to respond until they have seen a certain amount of motion; a difficulty not present for static images), making it difficult to compare across stimulus categories.

Houses were selected as comparison non-biological stimuli. Our house stimuli varied along dimensions such as feature shape, spacing, and texture; but shared a set number of features and a limited range of configurations (relationships between features), to increase the level of structural similarity between the stimuli (see Campbell & Tanaka, 2018). This allowed us to examine whether any of our DP participants demonstrated more general object agnosia, as opposed to a more specific impairment affecting only biological stimuli.

2. Materials and Methods

2.1. Participants

Forty adults with DP (22 female; age range = 18-66 years, $M = 47.6$, $SD = 14.6$) took part in this study. All had contacted our laboratory complaining of severe everyday difficulties with face recognition: these were confirmed via diagnostic protocols that are adhered to by most laboratories in the field (see Dalrymple & Palermo, 2016; Murray, Hills, Bennetts & Bate, 2018). In brief, all individuals performed significantly below published age-matched control cut-offs on the Cambridge Face Memory Test (CFMT; Duchaine & Nakayama, 2006; for cut-offs see Bowles et al., 2009) and a famous faces test (Bate, Adams, Bennetts & Line, in press). Each case's scores on these tests are presented as supplementary information (see SM1), in addition to individual scores on the upright trials of the Cambridge Face Perception Test (CFPT; Duchaine et al., 2007). Because face perception can sometimes be preserved in DP, CFPT scores are typically not used as an absolute diagnostic criterion (Bate & Tree, 2017; Dalrymple & Palermo, 2016), and are not regarded as such in the current study.

No individual reported a history of socio-emotional, psychiatric or neurological disorder. Concurrent socio-emotional disorder was also excluded using the Autism Quotient (Baron-Cohen, Hoekstra, Knickmeyer & Wheelwright, 2006), and cognitive decline in those aged 65+ (using the Mini Mental State Examination: Folstein, Folstein, & McHugh, 1975). Participants were screened for lower-level perceptual impairments: basic visual acuity was assessed using a standard Snellen letter chart (3 m), the Hamilton-Veale contrast sensitivity test, and four sub-tests of the Birmingham Object Recognition Battery (Line Match, Size Match, Orientation Match; Position of the Gap Match; Humphreys & Riddoch, 1993). No participant showed any impairment.

A total of 60 control participants (30 male) also took part in this study, IQ-matched to the DP sample (using the Wechsler Test of Adult Reading, Holdnack, 2001). Because of the

varied age in the DP sample, controls were recruited according to two separate age-groups, each containing 30 (15 female) participants. As previous work has indicated small improvements in face recognition until the age of 30 (Germine, Duchaine & Nakayama, 2011), followed by consistency until the age of 50 (e.g. Bowles et al., 2009), the younger group were aged 20-49 years ($M = 32.9$, $SD = 9.1$), and the older group 50-66 years ($M = 57.0$, $SD = 4.6$). Controls were recruited from the departmental participant pool, and received a small financial payment in exchange for their time. They underwent the same perceptual and socio-emotional screening procedures as described for the DP group, and no individual reported everyday difficulties in face recognition. Ethical approval for this experiment was granted by the institutional Ethics Committee, and all participants provided informed consent according to the Declaration of Helsinki.

2.2. Materials and procedure

A sequential same/different face and object (hand/houses) matching task, that displayed stimuli in upright and inverted conditions, was created within our laboratory and has been used in our previous work (Bobak et al., 2016). Existing analyses on control data found a clear inversion effect for faces but not hands or houses (Bobak et al., 2016), indicating the test is suitable for the assessment of differences in the processing of faces versus objects. Further, as the task restricts the length of time that initial images are displayed, DPs were unlikely to use laboured compensatory strategies to achieve correct responses. Nevertheless, both accuracy and reaction time were monitored (see below).

Test trials consisted of two sequentially presented images (see Figure 1) – the initial study image was displayed for 250 msec, and the second test image was displayed until the participant responded. The images were separated by a 1000 msec ISI (a fixation cross displayed in the centre of the screen). In the face condition, the study image showed a face

from a frontal viewpoint and the test image from a 30-45° angle. Faces were drawn from the Cambridge Face Memory Test-Australian (McKone et al., 2011) and the Bosphorous Face Database (Savran et al., 2008), and were edited to remove external features. Images were paired so that the target and distractor images for each test face were taken from the same database (to minimise differences in contrast and luminance), and presented the faces at the same angle. Houses were created using the software Realtime Landscaping Plus (Idea Spectrum Inc., 2012). Each house contained the same number of features (three sets of windows and a door), placed onto a constant background texture. The shape and exact location of the features, the background (building) texture, and the overall shape of the house varied throughout the set. As in the face condition, the study and test images presented the houses from two different viewpoints (frontal and 15° profile). To prevent matching based on low-level image characteristics such as the background colour of the houses (which was varied slightly throughout the stimulus set), the brightness of the test images was reduced by 30% using Adobe Photoshop. Hand images were extracted from the Bosphorus Hand Database (Dutagaci, Yoruk, & Sankur, 2008), and showed the palm and fingers of a hand. Images were chosen to exclude rings, watches, cuffs or other identifying features. Study and test images showed the hands in two different positions (e.g. fingers slightly splayed and fingers together), with the fingers pointing upwards (upright condition) or downwards (inverted condition). To prevent low-level image matching, the test images were processed further in Adobe Photoshop – the brightness was reduced by 30%, and a mosaic filter was applied with a cell size of 3 squares (resulting in a slightly pixelated image). Across all conditions, the stimuli were resized to measure approximately 8cm across when displayed on-screen, subtending roughly 10 degrees of visual angle when participants were seated at a comfortable distance from the monitor (50-60cm).

Each category contained 32 pairs of images (16 same identities, 16 different identities). All pairs were presented twice upright and twice inverted. Trials were blocked by stimulus type, with upright and inverted trials presented randomly within each stimulus type. The order of blocks was randomised between participants, and they were required to indicate whether the two images showed the same or different faces or objects by pressing the *z* and *m* keys on the keyboard (assignment of keys was counterbalanced between participants). Participants were instructed to respond as quickly and as accurately as possible.

< *Insert Figure 1* >

2.3. Statistical analyses

Because this task contained matching and mis-matching trials, we initially calculated the proportion of hits (correctly identifying that two images matched in identity) and correct rejections (correctly identifying that two images do not match in identity) that each participant made in each condition. We also went beyond existing work by examining accuracy independently of response bias, computing signal detection theory (SDT) measures of sensitivity (d') and bias (c) for each participant. d' incorporates information from hits and false positives to create a measure of sensitivity that is free from the influence of response bias (Macmillan & Creelman, 2005). A score of 0 indicates chance performance: values for the current test can range from -4.59 (consistently incorrect responding) to 4.59 (perfect accuracy). The measure c is used as an indicator of response bias (i.e. whether the participant has a tendency to say that the images do or do not match; MacMillan & Creelman, 2005). A score of 0 indicates a neutral response criterion, whereas a positive score indicates conservative responding (a tendency to indicate that a stimuli were not the same) and a negative score indicates more liberal responding (a tendency to indicate that the stimuli were the same). Prior to calculation of the SDT measures, extreme hit and correct rejection scores

(1 and 0) were adjusted by replacing them with $(n - 0.5)/n$ and $0.5/n$ respectively (when n indicates number of possible hits or false alarms; Stanislaw & Todorov, 1999). We also calculated mean response latencies for correct responses in each condition. Any reaction times that exceeded three SDs from each participant's mean response latency were excluded.

Overall patterns of performance in the control participants were initially carried out to assess the calibration of each condition was equal in difficulty. Subsequently, we compared DP to control performance in group-based analyses. Our remaining analyses focused solely on the DP group, beginning with a factor analysis on the overall data. Next, we took a categorical case-by-case approach to partitioning the data. Because it is well known that DP is heterogenous in its presentation, we expected a varying pattern of performance, even in the upright faces condition. Deficits in face perception are only sometimes observed in DP (e.g. Chatterjee & Nakayama, 2012; Palermo et al., 2011), and given the nature of our task (i.e. a sequential matching task), it is possible that some individuals may be able to achieve typical patterns of performance. However, as typical accuracy scores are sometimes associated with atypically long response latencies (see discussion above), we wanted to be conservative in identifying any potential cases of preserved performance. Because face inversion effects are often used as an indicator of typical face-processing strategies (Farah, Tanaka & Drain, 1995; Yin, 1969), and support for domain-specificity comes from findings of larger inversion effects for faces than objects (Maurer, Le Grand & Mondloch, 2002; Yin, 1969), our categorization criteria for preserved performance in the faces condition therefore required a typical face inversion effect, in addition to typical accuracy and reaction time.

Crawford and Garthwaite's (2005) Bayesian Standardized Difference Test (SDT) was used to estimate whether each individual's standardized difference between face and object performance differed from the standardized difference observed in the control sample. Significant differences were sub-divided into strong (where the participant displays impaired

scores on both tests) versus classic (where one score is impaired and one is intact) dissociations (Shallice, 1988). This is theoretically important: evidence of the latter suggests a qualitative difference between face and object processing (i.e. evidence of domain-specificity), whereas the former suggests only a quantitative difference, implying a reliance on related mechanisms and/or an association between impairments that affect different stimulus categories (Gerlach et al., 2018).

3. Results

3.1. Control patterns of performance

Initially, we examined overall patterns of performance in the control group only, to determine whether the face and object matching conditions were appropriately matched for difficulty. Subsequently, we examined overall patterns of performance in the two participant groups, to ensure there were no unusual patterns of results which might affect our later analyses (e.g. unusual inversion effects; extreme differences in bias across conditions). For brevity, only significant interactions and main effects are reported (all $ps > .05$ for non-significant results). The Huynh-Feldt correction is applied where relevant, and multiple comparisons are Bonferroni corrected.

The d' and reaction time data for the control group was entered into a 3 (stimulus category: faces, hands, houses) x 2 (orientation: upright, inverted) repeated measures ANCOVA, with participant age as a covariate. The ANCOVA on d' revealed that the main effect of stimulus was significant, $F(2,116) = 98.72, p < .0005, \eta^2 = .630$, as was the main effect of orientation, $F(1,58) = 11.55, p < .0005, \eta^2 = .571$, and the interaction between stimulus and orientation, $F(1.90,110.01) = 28.18, p < .0005, \eta^2 = .33$. Pairwise comparisons indicated that houses were discriminated significantly better than hands and faces, in both the upright and inverted conditions; hands were discriminated also discriminated better than

faces in the inverted condition (all $ps < .0005$). There was no significant difference between faces and hands in the upright condition, $p > .1$.

The ANCOVA on reaction time revealed that the main effect of orientation was significant, $F(1,58) = 15.29, p < .0005, \eta^2 = .209$, as was the interaction between stimulus and orientation, $F(1.74,101.10) = 13.38, p < .0005, \eta^2 = .187$. The main effect of stimulus was not significant, $F(2,116) = 1.62, p = .203$. Pairwise comparisons indicated that there were no significant differences between reaction times to upright stimuli; however, inverted faces were matched significantly slower than inverted hands or houses, $p's < .02$.

From these analyses, it is apparent that control participants were significantly better at matching the house stimuli, compared to the faces and hand stimuli. Consequently, we conducted a materials analysis to equate the difficulty levels of the different stimulus types. We used a median split based on accuracy to separate the upright house trials into two groups (separately for “same” and “different” trials), and those in the top half were removed from the analysis. The matched trials (i.e., those using the same stimuli) were removed from the inverted condition. This allowed us to preserve the match between upright and inverted stimuli, as well as the balance of same and different trials across conditions. Given that each trial was repeated twice in the experiment, this resulted in 16 trials per condition being entered into the analysis for the house stimuli. As the face and hand stimuli were well-matched for difficulty in the upright condition, they did not undergo any adjustment. Subsequent to the adjustment, follow-up analyses revealed that there was no longer a significant difference in performance between upright faces, houses, and hands in the control group (all $p's > 0.1$).

3.2. Group-based analyses: DPs versus controls

For d' , c and reaction time, data were entered into a 2 (participant group: DP, control) x 3 (stimulus category: faces, hands, houses) x 2 (orientation: upright, inverted) mixed factorial ANCOVA, with participant age as a covariate. A further analysis also included the proportion of hits and correct rejections (trial type) as an additional factor.

d' (sensitivity): The ANCOVA revealed significant two-way interactions for stimulus and participant group, $F(1.90, 184.72) = 6.22, p = .003, \eta p^2 = .060$, orientation and participant group, $F(1, 97) = 3.93, p = .050, \eta p^2 = .039$; and orientation and stimulus, $F(2, 194) = 27.44, p < .0005, \eta p^2 = .221$ (see Figure 2). The three-way interaction was not significant. Follow-up analyses confirmed that DPs performed significantly worse than controls when matching faces ($p < .0005$), but not hands or houses ($ps > .05$). Averaged across stimuli, DPs also performed significantly worse than controls in upright trials ($p = .001$), but not inverted trials ($p = .098$). Across groups, there was a significant inversion effect for faces ($p < .0005$) and hands ($p = .010$), but not houses ($p = .914$).

These interactions superseded main effects of participant group (where controls outperformed DPs: $F(1, 97) = 7.46, p = .008, \eta p^2 = .071$), orientation (where upright stimuli were recognized more accurately than inverted: $F(1, 97) = 47.88, p < .0005, \eta p^2 = .330$), and stimulus (houses were recognized more accurately than faces or hands, and hands more accurately than faces; all $ps < .001$), $F(1.90, 184.72) = 66.61, p < .0005, \eta p^2 = .407$.

< Insert Figure 2 >

Hits and correct rejections: The four-way interaction was not significant, nor were any of the three-way interactions involving participant group (see Figure 3). There was a three-way interaction between stimulus, orientation, and trial type, $F(2, 194) = 13.82, p < .0005, \eta p^2 = .125$; reflecting an inversion effect for correct rejections of faces ($p < .0005$), but not houses or hands ($ps > .1$); but no inversion effect for hits for any stimulus ($ps > .15$).

< Insert Figure 3 >

All of the two-way interactions involving stimulus were significant. The interaction between stimulus and orientation, $F(2,194) = 27.85, p < .0005, \eta^2 = .223$, reflected a significant inversion effect for faces and hands (p 's $< .05$) but not houses ($p = .891$). The interaction between stimulus and trial type, $F(1.89,182.92) = 38.26, p < .0005, \eta^2 = .283$, reflected the fact that there were significant differences between stimulus categories in the upright trials (faces less than hands and houses, hands less than houses, all p 's $< .01$), but not in the inverted trials (p 's $> .25$). The interaction between stimulus and participant group, $F(1.90,184.29) = 7.00, p = .001, \eta^2 = .067$, reflected a significant difference between DPs and controls in the faces condition ($p < .0005$, DPs $<$ controls), but not the houses or hands conditions (p 's $> .05$).

There was also a significant interaction between orientation and trial type, $F(1,97) = 7.44, p = .008, \eta^2 = .071$: the inversion effect for hits was not significant ($p = .059$), whereas it was for CRs ($p < .0005$). The interaction between orientation and participant group, $F(1,97) = 7.50, p = .007, \eta^2 = .072$ also reached significance, reflecting a significant difference between controls and DPs in upright trials ($p < .0005$) but not inverted trials ($p = 0.91$). Unsurprisingly, there were main effects of participant group and orientation, $F(1,97) = 8.40, p = .005, \eta^2 = .080$, and $F(1,97) = 61.54, p < .0005, \eta^2 = .388$, respectively. A main effect of stimulus, $F(1.90,184.29) = 65.65, p < .0005, \eta^2 = .404$, was underpinned by better performance for houses and hands compared to faces ($ps < .0005$), and better performance for houses than hands ($p = .004$). A main effect of trial type, $F(1,97) = 24.14, p < .0005, \eta^2 = .199$ reflected better performance in “same” trials (hits) than “different” trials (CRs).

c (bias): The three-way interaction was non-significant, as was the two-way interaction between participant group and stimulus. There was a significant interaction between participant group and orientation, $F(1,97) = 4.26, p = .042, \eta^2 = .042$. Control participants showed a more conservative response bias for upright than inverted stimuli ($p <$

.0005), whereas DPs showed no effect of inversion on bias ($p = .736$). There was also a significant interaction between stimulus and orientation, $F(2,194) = 14.92, p < .0005, \eta^2 = .133$; there was a significant difference in response bias between upright and inverted faces ($p < .0005$), with upright faces eliciting more conservative responses than inverted faces; however, there was no significant difference in response bias for upright and inverted hands and houses (p 's $> .3$). In addition to the interactions, there were main effects of orientation (responses for upright stimuli were more conservative than for inverted: $F(1,97) = 6.74, p = .011, \eta^2 = .065$) and stimulus, $F(1.90, 18.37) = 40.90, p < .0005, \eta^2 = .297$: responses were more conservative for faces than hands or houses (p s $< .0005$), but no difference between the latter ($p = .390$). The main effect of participant group was not significant.

Reaction time: The three-way interaction was not significant; nor were the two-way interactions involving participant group, or the main effect of stimulus (see Figure 4). There was a significant main effect of orientation (upright stimuli were matched faster than inverted stimuli: $F(1,97) = 6.76, p = .011, \eta^2 = .065$) and an interaction between stimulus and orientation, $F(2,194) = 9.50, p < .0005, \eta^2 = .089$. Pairwise comparisons revealed that there was a significant inversion effect for faces ($p < .0005$), but not hands or houses (p 's $> .2$). The main effect of participant group was also significant, $F(1, 97) = 9.34, p = .003, \eta^2 = .857$, reflecting slower reaction times for DPs compared to controls overall.

< Insert Figure 4 >

3.3. DPS: Overall patterns of performance

Despite our limited sample size ($N = 40$), the DP group's d' scores were entered into a principal components analysis (PCA). Initial eigenvalues indicated that the first two factors explained 48.48% and 17.35% of the variance, and the remaining four factors had eigenvalues that were less than one. Solutions for two, three, four and five factors were each

examined using varimax and oblimin rotations of the factor loading matrix. The three factor varimax solution (which explained 78.46% of the variance) was preferred, as it offered the best defined factor structure (see Table 1). The first factor had high loadings from upright and inverted performance on hands and houses. The second factor had high loadings from the inverted faces and upright and inverted hands conditions. The final factor had a very high loading from the upright faces condition, and a small loading from inverted hands. No significant correlations were observed between upright faces and any other condition, whereas moderate-to-strong correlations were observed between the four object conditions (see Table 2).

< Insert Tables 1 and 2 >

3.4. DPs: Face matching

Because we adopted a sequential matching task, it is unclear whether this task draws more heavily on perceptual or short-term memory mechanisms. To explore this issue, we carried out a PCA (using Varimax rotation) on the DP group's CFMT and CFPT scores (upright trials only, see SM1; note that CFPT scores were transformed into proportion correct, rather than raw number of errors), and their d' scores from the upright faces condition of the matching task. Two factors emerged that had eigenvalues that were greater than one. The first explained 40.81% of the variance, and had high loadings from the CFPT and the matching task (see Table 3). The second factor explained a further 36.01% of the variance, and had a very high loading from the CFMT.

< Insert Table 3 >

We then took a categorical approach to partitioning performance according to intact and impaired face matching performance. Seventeen DPs (eight male; M age = 33.3 years, SD = 10.4) were compared to the younger control group, and 23 (13 female; M age = 58.1

years, $SD = 5.0$) to the older group (see Table 4). Twenty-five out of the 40 DPs (62.5%) displayed intact face matching performance, judged by performance that was within 1.96 SDs of the control mean for both d' and reaction time in the upright faces condition, and the d' and reaction time face inversion effects. Of the 15 DPs who displayed impaired performance, eight only showed atypical d' scores, four only showed atypical reaction times, and two were impaired on both measures. One further participant achieved an impaired d' score, and their reaction times were significantly quicker than those of control participants.

< *Insert Table 4* >

3.5. DPs: Domain-Specificity

We then examined the proportion of DPs who showed domain-specificity within the two face matching groups (i.e. impaired and preserved). The same criteria were used to judge intact object matching skills as were used for face matching (see above): performance was required to be within 1.96 SDs of the relevant control mean on both d' and reaction time, in all object categories. We included both the inverted as well as the upright conditions in these criteria as it is debateable which orientation is truly “upright” for hands; as we subsequently did not want to imbalance the number of criterion conditions for hands versus houses, we also included the inverted houses condition. Note that we did not look at inversion effects for hands and houses as they are typically absent or reduced for objects (and were absent in our overall ANCOVAs).

Of the 25 DPs who showed typical face matching performance, eight achieved a z score that was within the impaired range in at least one object category (two were impaired across both categories, four only at houses, and two only at hands). SDTs confirmed classical dissociations (intact face but impaired object matching) in three individuals, all affecting only the matching of hands (see “reverse” dissociations in Table 5).

The remaining 15 DPs were within the “impaired face matching” category. Nine demonstrated intact object matching in all conditions. SDTs revealed significant dissociations between face and object matching in nine of the 15 DPs (see Table 5): six displayed classic dissociations (impaired face matching but typical object matching) and three displayed strong dissociations (impaired scores in both conditions). Of the latter three, one had impairments that affected both hands and houses, whereas two had impairments that were restricted to hands (see Table 5 for SDT results; see Figure 5 for Z-scores for all of the DPs who showed significant dissociations).

< Insert Table 5 and Figure 5 >

The proportion of DPs that fell into the intact and impaired face matching categories is displayed in Figure 6 as a function of confirmed dissociations with object matching. A Chi-Square analysis indicated a significant difference in the pattern of dissociation observed in each group, $X^2_2 = 10.29, p = .001$. Unsurprisingly, when face matching skills were spared, object matching skills were much more likely (7.3 times) to be unaffected than impaired. More interestingly, when face matching skills were impaired, object matching skills were dissociated in nine out of 15 individuals (including six classical dissociations).

< Insert Figure 6 >

3.6. DPs: Hands versus houses

Finally, we examined the specific patterns of object recognition deficits displayed by DPs. Fourteen individuals (eight with intact face matching) achieved at least one z score that was below 1.96 SDs of the control mean across the object conditions. Both hands and houses were affected in five DPs (three of these also showed face matching impairments), just hands in four (two with a face matching impairment), and just houses in five (one with impaired face matching skills). A Fishers Exact test indicated that there was not a significant

difference in the distribution of object impairments for DPs with intact and impaired face matching skills ($p = .577$). In other words, there was no significant association between the presence or absence of face perception deficits and the presence of a specific pattern of object perception deficits.

When examining the group of DPs with confirmed dissociations between face and object matching (12 individuals, see Table 5), six of the nine participants that presented with impaired face matching (listed as “Classic” and “Strong” dissociations in Table 5) displayed a dissociation for faces versus both hands and houses. Two individuals showed dissociations (one classic) between face and house matching, and one showed a classic dissociation between face and hand matching. The three remaining individuals showed dissociations between intact face matching and impaired object matching (all hands only; denoted as “Reverse” in Table 5).

We then screened the entire DP group for dissociations between performance in the hands and houses conditions, using SDTs. We compared performance in the upright houses condition to those of both upright and inverted hands (we included both hands condition given there is not a clear “upright” condition for this category; see Table 6). One individual (DP15), who had showed strong dissociations between face matching and both categories of objects, also showed a classic dissociation between impaired hand matching and intact house matching. One further DP (DP23) who was impaired at face matching, but did not dissociate from object matching, showed a classic dissociation between impaired house matching and intact hand matching. An additional participant (DP36) who was not impaired at face matching showed a classic dissociation between house and hand matching (hands impaired). Strong dissociations between house and hand matching were observed in two additional participants (DP29 and DP31) who did not present with face matching impairments.

< *Insert Table 6* >

4. Discussion

This investigation set out to examine the domain-specificity of face matching skills in a large sample of DP participants. While significant correlations emerged between all of the four object categories, no condition correlated with performance in the upright faces condition, and a PCA revealed separate factors for upright face perception and object perception.

Further, a categorical analysis demonstrated that, when face matching was impaired, object matching skills were classically dissociated in six out of 15 individuals. There was evidence of dissociation between face and object matching in one third of the 40 DP cases (including individuals with both intact and impaired face matching), and in many cases (20% of the overall group), this dissociation appeared across multiple object categories.

Importantly, evidence about domain-specificity has emerged in both group and case-by-case analyses. At the group-level, the performance of the DP group on the upright faces condition did not correlate with any other condition, despite moderate-to-high correlations between the four object conditions. This pattern of findings indicates that face and object matching deficits do not tend to associate according to their severity, and is backed up by evidence of statistical dissociations in some of the DP cases. Unsurprisingly, we found that, when face matching skills are intact, object matching skills also tend to be unaffected. However, when face matching skills were impaired, classical dissociations were noted in just over one third of the participants. While this is a low proportion of our overall sample and indicates that face-specific cases of DP are relatively rare, it is a higher proportion to the 20% figure reported by Geskin and Behrmann (2018). Further, it has the potential to substantially increase if any of our DPs who showed typical face and object matching performance go on to show domain-specific impairments in memory paradigms. Importantly, the larger figure in our study emerged despite the fact that Geskin and Behrmann drew conclusions across studies that used a wide variety of paradigms and initial inclusion criteria. Further, they used

z scores alone to deem intact or impaired performance, whereas an advantage of the current work is that we confirmed dissociations via conservative SDTs that account for the correlation of performance in control participants. Thus, while the current findings more rigorously assess domain-specificity in DP, they return greater support for domain-specificity than the more liberal criteria used by Geskin and Behrmann.

Placing proportions aside, the patterns of dissociation are of greater theoretical interest, particularly in the six individuals who demonstrated classical dissociations in their face matching impairments. Importantly, we are confident that these individuals genuinely fulfil the criteria for DP due to our stringent inclusion criteria. Further, our criteria for deeming typical face and object matching was mostly identical (although we also required evidence of a normal face inversion effect for the former): participants had to display typical performance on both d' and reaction time measures. While the individuals in the “impaired face matching group” achieved a range of z scores in the upright faces condition, four scored at least three SDs lower than the relevant control mean on d' , and four on reaction time. Thus, we can be confident that face matching was not just borderline-impaired in these individuals. Conversely, some of these DPs consistently achieved positive z scores across the object conditions. While two individuals only displayed the dissociation for one object condition, evidence was more consistent in the remaining four. This indicates a potential qualitative difference in processing between faces and other body parts, and between faces and other objects which show a high degree of structural similarity. The finding is particularly notable since the object categories were chosen to maximise the potential overlap between face and object processing. As such, our results provide convincing statistical support (backed up by careful methodology) for domain-specificity in at least four individuals. Pertinently, evidence for the double dissociation was also observed in three DPs who showed intact face matching but impaired object matching. While this finding is surprising, it is likely that individual

differences exist for all kinds of biological categories of stimuli, and category-specific 'agnosia' may be found in a small percentage of people. Nevertheless, the dissociation only emerged in one or two comparisons in each individual, and would need replication before further comment.

There is also evidence to suggest more complex patterns of object recognition skills in DP. The results from the PCA indicated that hand and house perception loaded onto the same factor, suggesting some shared processing mechanisms across the two object categories. However, the DPs showed a relatively heterogeneous pattern of object matching impairments (a mix of hand only, house only, and multiple object deficits), which argues against a single common deficit underlying all cases of DP (e.g. Geskin & Behrmann, 2018). In support of this, some participants also demonstrated a dissociation between hand and house perception. Notably, this dissociation was seen in participants with and without face matching deficits, and in the presence and absence of dissociations between face and object recognition, suggesting it is not necessarily linked with the core face recognition deficit in DP. This is in line with recent work that has argued against a simple, single mechanism explanation for DP: for instance, Biotti, Gray and Cook (2017) found little relationship between the perception of cars and bodies in their DP participants, although both correlated with performance in the face condition. The authors suggest that such variations in object recognition skills may indicate co-occurrence of different developmental conditions, rather than damage to a common underpinning mechanism. The extent to which object categories may be partitioned under this account remains to be seen, although other studies have presented evidence of varied performance for different non-biological classes of object (e.g. Dalrymple, Elison & Duchaine, 2017). Alternatively, it remains possible that DPs may have a domain-general deficit that impairs perception of some object categories more than others (e.g. impaired processing of low-spatial frequencies, configurations, or curvature), and this has not been

tapped by the object categories used in the current or previous studies. Thus, future work should employ further classes of biological and non-biological objects to explore this point, although it remains imperative that selection of such categories remains theoretically-driven and appropriately comparable to faces.

A secondary point of interest from this investigation concerns the wide variation in performance on the upright faces condition. While it is unsurprising to see this heterogeneity, it is difficult to interpret what this variance actually represents. Indeed, the matching paradigm is likely to draw on both short-term memory and perceptual mechanisms, although it is notable that performance in the upright faces condition loaded heavily on the same factor as the CFPT in our PCA. Further, our finding that 62.5% of the 40 DPs met criteria for “typical” face matching performance is nearly identical to the proportion of DPs (from a sample of 16) reported to have intact face perception skills by Dalrymple and colleagues (2014). In line with Dalrymple et al.’s (2014) work, we used a conservative criterion for classifying DPs into the “intact” category: not only did we take performance that was within the typical range on both SDT and reaction time measures, but we also used the face inversion effect to indicate typical face-specific processing. In addition, our use of SDT enabled us to be confident that any individual scored within the typical range because they genuinely recognised the stimuli, and not as a result of response bias. Finally, given that our inclusion protocols ensured that all our DP participants had typical lower-level vision, the differing patterns of performance on the matching paradigm suggests that higher-order perceptual processes are being tapped, without placing demands on long-term face memory (given this is known to be impaired in the entire DP group).

While these findings suggest a distinction between two different types of DP, further exploration is needed to ascertain whether this maps exactly onto the apperceptive/associative sub-division proposed for AP (e.g. De Renzi, Faglioni, Grossi, & Nichelli, 1991).

Nevertheless, the current findings are important, given most existing evidence supporting impaired (e.g. Bate & Cook, 2012; Chatterjee & Nakayama, 2012; Duchaine et al., 2007; Duchaine & Nakayama, 2006; Palermo et al., 2011; Yovel & Duchaine, 2006) or intact (Behrmann et al., 2005; Chatterjee & Nakayama, 2012; McKone et al., 2011; Palermo et al., 2011) face perception skills in DP have failed to take both accuracy and reaction time into account (but see Dalrymple, Garrido & Duchaine, 2014; Ulrich et al., 2017). Furthermore, no known study has examined the face-specificity of different patterns of perceptual skills in DP, and to date, most of the debates around domain-specificity have not distinguished between mnemonic and perceptual deficits (see Barton, 2018). This is an important theoretical issue as the face-specificity of apperceptive compared to associative prosopagnosia in AP is unclear, and some authors believe that the former is indicative of a more general form of visual agnosia, while associative prosopagnosia represents a category-specific impairment of semantic memory (De Renzi et al., 1991; Benton & Tranel, 1993). Thus, the pattern of dissociations reported here presents novel evidence supporting the existence of different functional subtypes in DP.

In sum, the work presented here provides timely new evidence about domain-specificity in DP. As a recent review and associated commentaries (see the editorial by Susilo, 2018) called all existing evidence on this issue into potential disregard, the careful methodology and analysis adopted here present a new starting point for the field. Importantly, findings suggest domain-specificity in six out of 15 DPs who were impaired at face matching. Admittedly, this is a low proportion of the overall sample, and our current evidence does not suggest domain-specificity in the majority of DP cases. Thus, future work should consider whether these figures hold across different measures of face and object perception, and whether the evidence for domain-specificity extends to memory paradigms. Future screening and remediation programmes may need to account for these emerging subtypes of DP.

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Supplementary Materials

All data supporting this manuscript are available as supplementary materials. Background diagnostic data for all participants with DP can be found in SM1. Raw data for control and DP participants for the experimental task is presented in SM2.

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Figure Captions

Figure 1: Sample stimuli from the faces, hands and houses conditions.

Figure 2: Estimated marginal means of d' scores for control and DP participants, controlling for age. Error bars represent ± 1 SEM.

Figure 3: Estimated marginal means of a) hits and b) correct rejections for control and DP participants, controlling for age. Error bars represent ± 1 SEM.

Figure 4: Estimated marginal means of reaction time for control and DP participants, controlling for age. Error bars represent ± 1 SEM.

Figure 5: Summary of standardized scores for all DPs who showed significant dissociations between face and object recognition. Top panels show standardized d' scores for DPs who showed a) classic dissociations; and b) strong (grey lines) and reverse (black lines) dissociations. Bottom panels show standardized RT for DPs who showed c) classic dissociations; and d) strong (grey lines) and reverse (black lines).

Figure 6: The proportion of DPs that fell into the intact and impaired face perception categories, as a function of confirmed dissociations with object perception.

Tables

Table 1: Varimax rotated component loadings for DPs' upright and inverted d' scores for faces, hands and houses (values $< .3$ are suppressed).

Component	1	2	3
Upright faces			.96
Inverted faces		.94	
Upright hands	.60	.59	
Inverted hands	.70	.31	.37
Upright houses	.88		
Inverted hands	.76		

Table 2: Correlation matrix for d' scores for DP participants for upright (U) and inverted (I) conditions.

	Faces U	Faces I	Hands U	Hands I	Houses U	Houses I
Faces U	1	.05	.17	.33	.29	.18
Faces I		1	.47*	.30	.17	.31
Hands U			1	.65**	.49**	.43*
Hands I				1	.64**	.38*
Houses U					1	.53**
Houses I						1

* $p < .006$; ** $p < .001$ (Holm's sequential Bonferroni correction applied)

Table 3: Varimax rotated component loadings for DPs' CFMT and upright CFPT scores, together with d' scores for the upright faces condition in the matching task (values $< .3$ are suppressed).

Component	1	2
CFMT		.92
CFPT	.75	.39
Upright faces matching	.81	

Table 4: Mean (SD) upright (U) and inverted (I) d' and reaction time (RT) performance for younger and older controls in each condition, with the inversion effect (IE) for faces.

	Faces			Hands		Houses	
	U	I	IE	U	I	U	I
Younger							
d'	2.14	1.37	0.77	2.10	1.96	2.36	2.20
	(0.37)	(0.64)	(0.61)	(0.52)	(0.48)	(0.61)	(0.52)
RT	960.55	1078.47	117.93	1011.71	1010.53	1017.62	1062.10
	(193.87)	(240.69)	(138.97)	(189.59)	(219.36)	(244.94)	(264.88)
Older							
d'	1.99	1.13	0.86	1.93	1.88	2.02	1.97
	(0.54)	(0.66)	(0.77)	(0.65)	(0.67)	(0.70)	(0.97)
RT	1122.71	1264.77	142.06	1113.44	1132.13	1226.13	1233.11
	(307.89)	(405.21)	(200.85)	(281.65)	(328.11)	(387.20)	(331.12)

Table 5: The results of Bayesian Standardized Difference Tests that confirmed dissociations between performance in the upright faces condition compared to the upright (U) and inverted (I) hands and houses conditions. Reported statistics represent t values; the estimated percentage of the control population exhibiting a difference more extreme than the individual is presented in parentheses.

	d'				Reaction Time			
	U hands	U houses	I hands	I houses	U hands	U houses	I hands	I houses
Classic:								
DP10	1.16 (12.82)	2.69 (0.58)*	2.48 (0.96)	1.64 (5.57)	2.06 (2.43)	3.03 (0.26)*	2.26 (1.57)	3.38 (0.10)*
DP12	2.57 (0.78)*	2.99 (0.28)*	2.55 (0.82)*	1.95 (3.02)	12.41 (0.01)**	12.78 (0.01)**	10.21 (0.01)**	12.78 (0.01)**
DP13	2.56 (0.79)*	2.47 (0.97)*	1.71 (4.86)	4.07 (0.02)**	1.03 (15.56)	0.39 (35.00)	0.90 (18.85)	0.35 (36.30)
DP17	2.90 (0.35)*	1.76 (4.43)	1.93 (3.20)	1.69 (5.13)	2.68 (0.60)*	2.00 (2.74)	2.19 (1.85)	1.95 (3.03)
DP32	2.89 (0.36)*	2.05 (2.50)*	3.48 (0.08)*	3.25 (0.15)*	0.17 (43.47)	0.71 (24.04)	0.43 (33.60)	0.06 (47.66)
DP35	4.30 (0.01)**	3.15 (0.19)*	3.42 (0.09)*	3.56 (0.07)**	0.34 (36.87)	0.87 (19.53)	0.15 (44.13)	0.58 (28.28)
Strong:								
DP07	0.72 (23.80)	0.15 (44.07)	0.29 (38.56)	0.93 (17.98)	2.22 (1.73)	2.67 (0.06)*	1.41 (8.46)	3.40 (0.10)*
DP15	2.53 (0.85)*	1.67 (5.33)	2.00 (2.76)	3.09 (0.22)*	2.45 (1.04)	0.73 (23.63)	2.06 (2.40)	0.53 (30.07)

DP25	1.59 (6.17)	0.41 (34.37)	1.32 (9.93)	0.09 (46.28)	8.10 (0.01)**	7.85 (0.01**)	9.40 (0.01)**	6.24 (0.01)**
Reverse:								
DP29	0.67 (25.56)	0.69 (24.66)	0.50 (31.18)	0.63 (26.79)	3.97 (0.02)**	2.41 (1.13)	0.14 (44.58)	0.49 (31.51)
DP31	0.13 (44.93)	1.58 (6.24)	0.89 (19.06)	1.61 (5.92)	6.52 (0.01)**	1.94 (3.09)	0.73 (23.50)	1.00 (16.32)
DP36	1.08 (14.39)	1.28 (10.56)	0.64 (26.35)	1.14 (13.27)	5.17 (0.01)**	0.45 (32.73)	3.73 (0.04)**	0.06 (47.74)

** $p < .001$; * $p < .05$ (Holm's sequential Bonferroni correction applied)

Table 6: The results of Bayesian Standardized Difference Tests that confirmed dissociations between performance in the upright houses condition compared to the upright and inverted hands conditions. Reported statistics represent t values; the estimated percentage of the control population exhibiting a difference more extreme than the individual is presented in parentheses.

	Face matching impaired	d'		RT	
		Upright hands	Inverted hands	Upright hands	Inverted hands
Classic:					
DP15	Yes	0.52 (30.45)	0.16 (43.88)	2.64 (0.66)*	2.49 (0.94)*
DP23	Yes	2.92 (0.33)*	2.52 (0.88)*	1.86 (3.63)	0.86 (19.75)
DP36	No	0.43 (33.42)	1.01 (16.13)	3.65 (0.05)**	2.62 (0.69)*
Strong:					
DP29	No	0.15 (44.16)	0.36 (36.14)	0.50 (31.13)	2.50 (0.91)*
DP31	No	1.89 (3.42)	1.12 (13.58)	3.12 (0.20)*	1.50 (7.18)

** $p < .001$; * $p < .05$ (Holm's sequential Bonferroni correction applied)

Figures

Figure 1

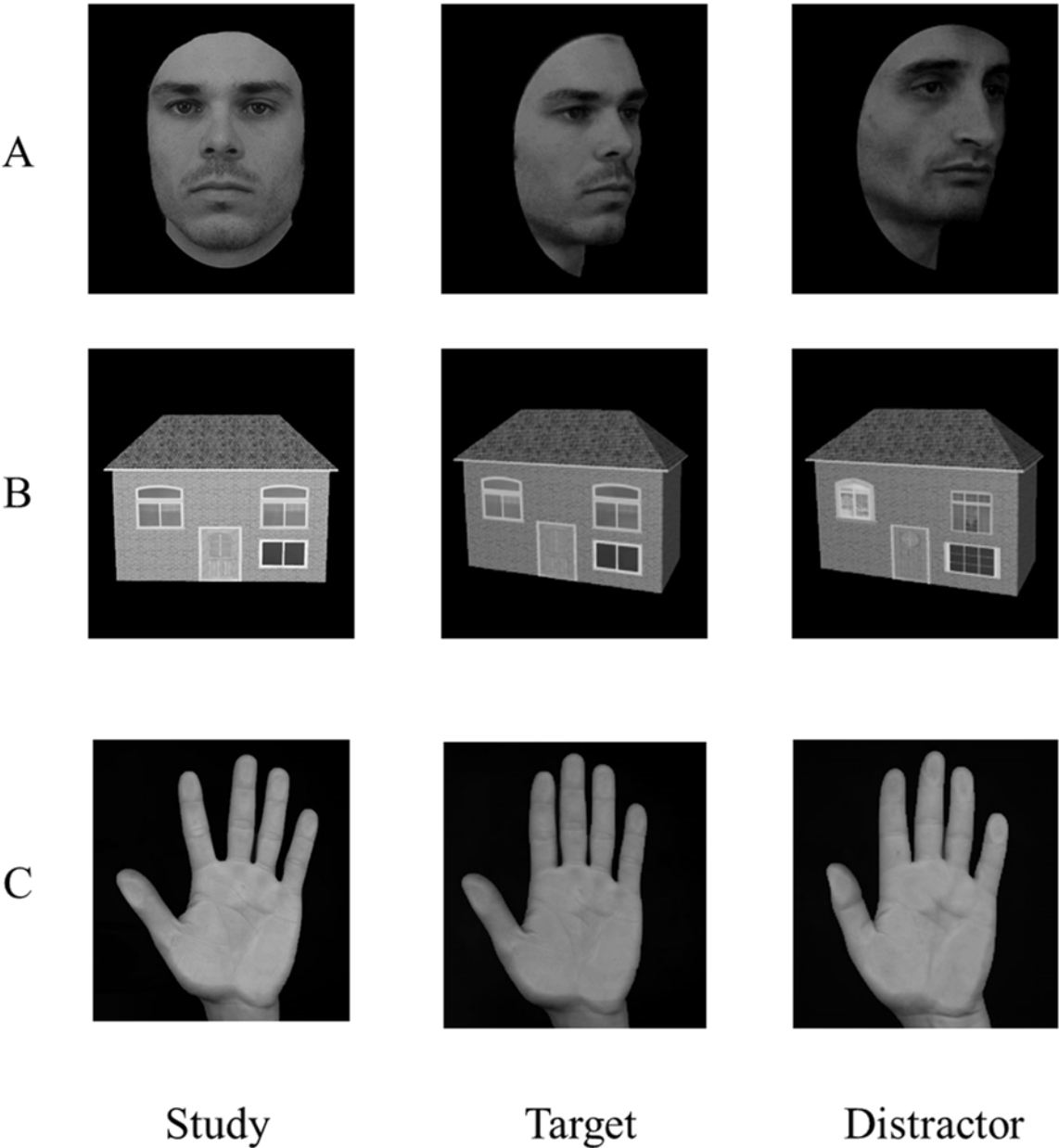


Figure 2

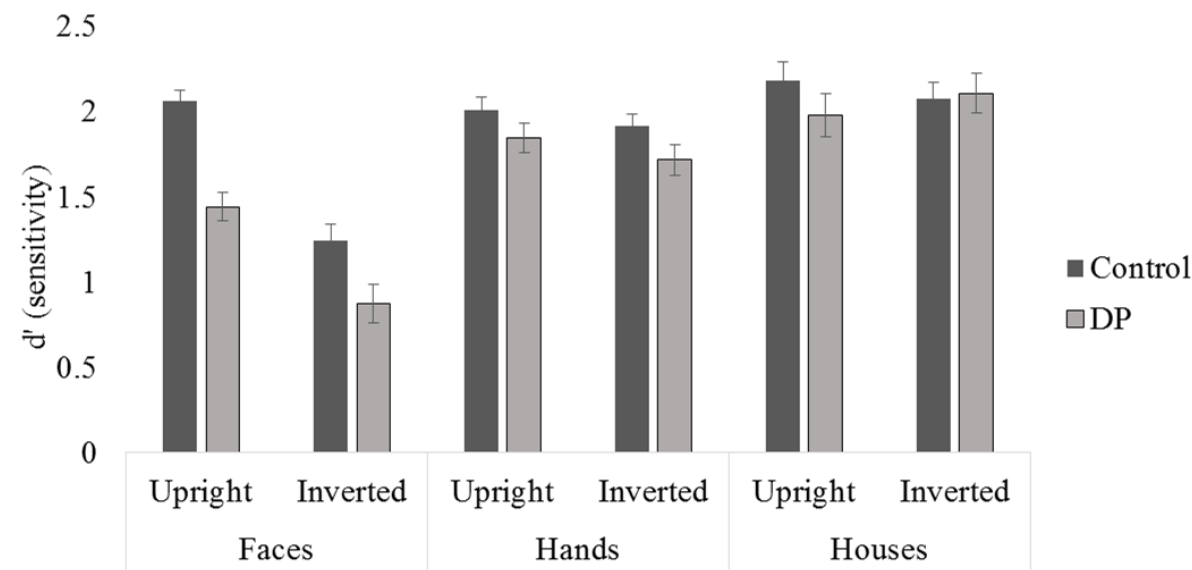


Figure 3

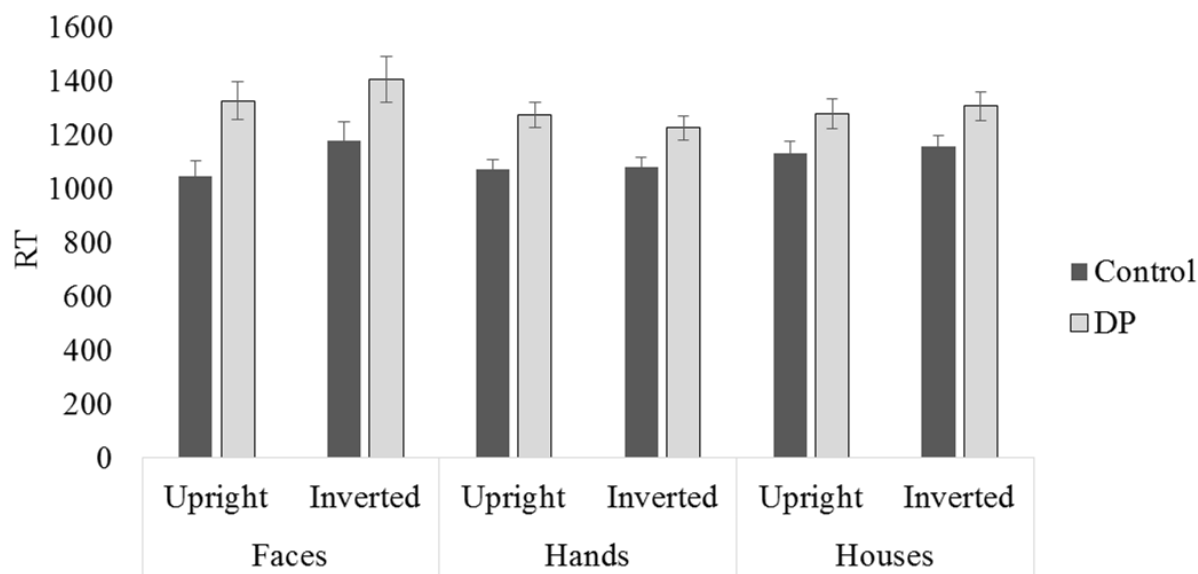


Figure 4

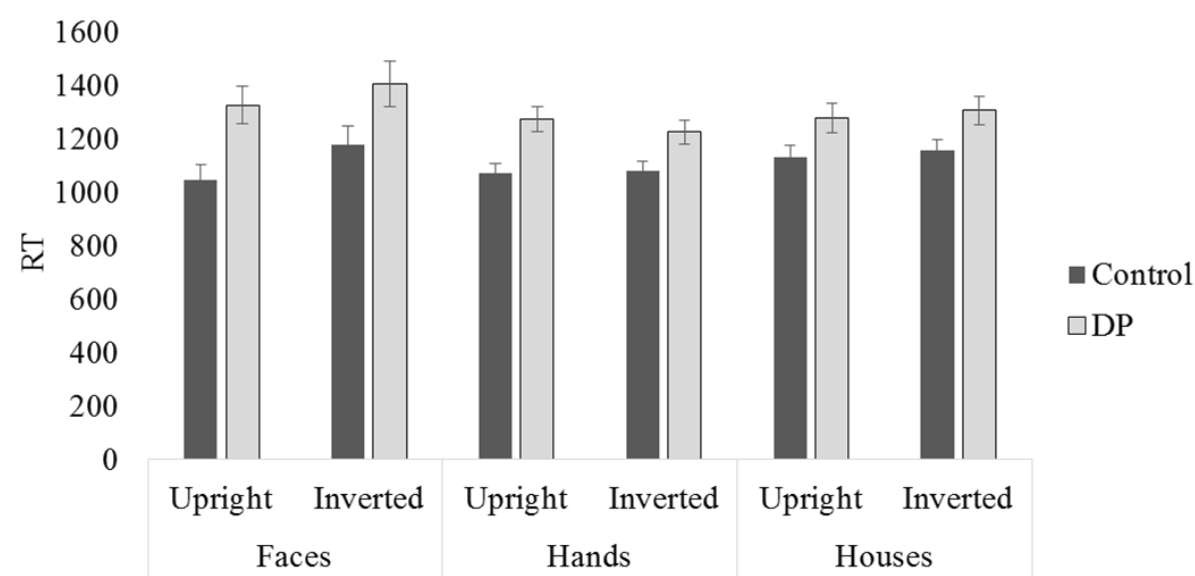


Figure 5

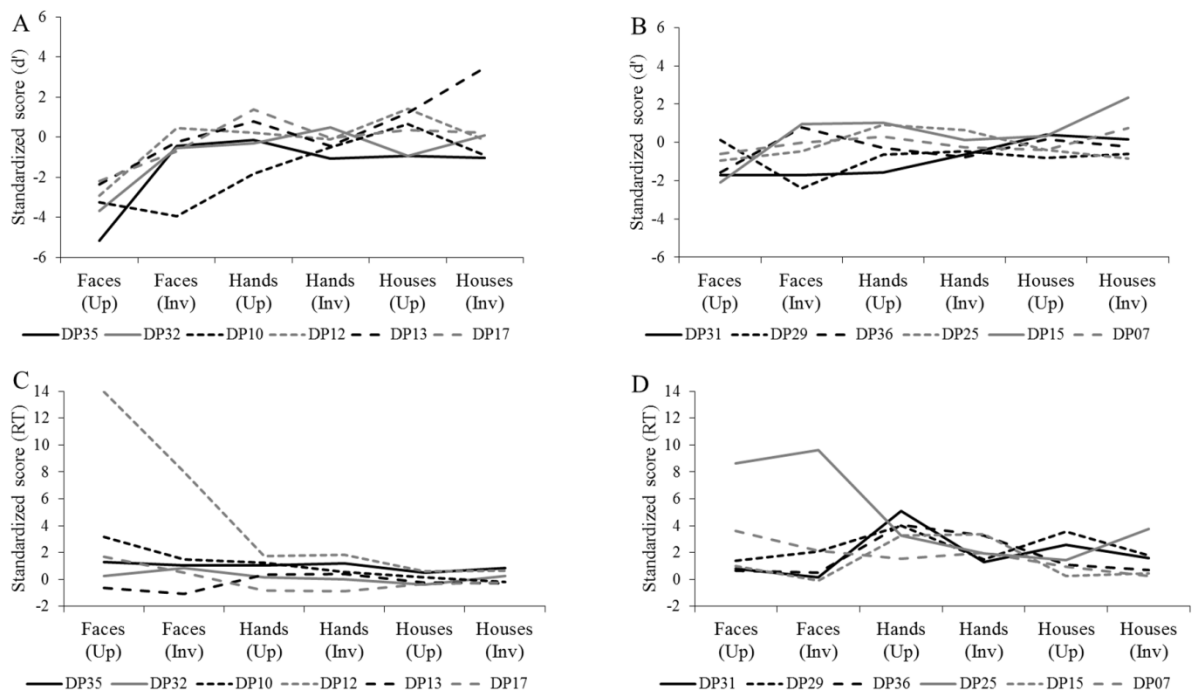


Figure 6

